Intraoperative Aneurysmal Subarachnoid Hemorrhage After Rupture of a Previously Undiagnosed Intracranial Aneurysm During Chiari Decompression: Case Report and Literature Review

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Abstract

Type 1 Chiari malformation occurs when caudal displacement of cerebellar tonsils below the level of foramen magnum obstructs the normal flow of cerebrospinal fluid (CSF) between the cranial and spinal spaces, a condition that often needs surgical decompression to restore normal CSF circulation. Abrupt changes in CSF flow dynamics after Chiari decompression can affect the intracranial CSF dynamics to the extent that a previously undiagnosed intracranial aneurysm remote from the site can rupture. The authors describe the development of intraoperative aneurysmal subarachnoid hemorrhage that occurred as a result of spontaneous rupture of a previously undiagnosed right distal posterior inferior cerebellar artery in a 57-year-old woman with type 1 Chiari malformation who was undergoing surgical decompression. The mechanism of the aneurysmal rupture appears to be related to the changes of CSF flow dynamics during surgical decompression. Normally, pressure equilibrium between the two sides of the aneurysmal wall prevents its rupture, but factors that significantly affect this equilibrium, such as systemic hypertension, can cause the aneurysm to rupture. The concept of spontaneous intraoperative rupture of intracranial aneurysm remote from the site of surgery has been described twice previously but under different scenarios. This scenario, to the best of our knowledge, has not been previously described.

Key Words: Chiari malformation type 1; craniovertebral decompression; subarachnoid hemorrhage; intraoperative complications; aneurysm; rupture

Running Title: Spontaneous intraoperative rupture of remote intracranial aneurysm

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Introduction

Type 1 Chiari malformation occurs when pathological caudal displacement of cerebellar tonsils below the foramen magnum obstructs cerebrospinal fluid (CSF) flow between the cranial and spinal subarachnoid spaces.[1,2] This acquired anomaly can be associated with spinal abnormalities such as syringohydromyelia, scoliosis, and hydrocephalus[1] and less commonly with cranial abnormalities. Physiologic studies have demonstrated abnormal CSF and blood dynamics at the craniovertebral junction (CVJ) in these patients.[1,2] We present a case of intraoperative subarachnoid hemorrhage (SAH) secondary to rupture of an undiagnosed intracranial aneurysm during suboccipital craniectomy for Chiari decompression. This event has not been reported previously, but intraoperative rupture of an unnoticed intracerebral aneurysm remote from the surgical site has been reported twice under different scenarios.[3,4] We consider possible reasons for this aneurysmal rupture by discussing CSF and cerebrovascular dynamics of patients with Chiari malformation type I.

Case Report

A 57-year-old woman presented with several-year history of positional tinnitus and suboccipital headaches that were precipitated by standing upright and alleviated with supine positioning. MRI of the brain revealed 8 mm of cerebellar tonsillar descent below the foramen magnum, consistent with Type I Chiari malformation (Fig. 1).

The patient underwent suboccipital craniectomy, C1 laminectomy, and duraplasty for Chiari decompression. Before incision, she was given 50 g of mannitol for cerebral relaxation. During bone removal at the foramen magnum, her blood pressure elevated acutely coincident with temporary loss of somatosensory evoked potentials from the upper and lower extremities,

and the dura became tense. No dural violation was noted. Upon dural opening, although the arachnoid was intact, there was massive filling of the subarachnoid space with bright red hemorrhage. The arachnoid layer was opened, and the subarachnoid space was copiously irrigated until the CSF was cleared of hemorrhage. No obvious bleeding source was identified, and the operation was finished uneventfully.

Immediate postoperative CT angiography revealed blood in the basal cisterns consistent with aneurysmal SAH, but no source of the hemorrhage (Fig. 2A). Formal cerebral angiogram demonstrated a slow filling of a left anterior inferior cerebellar artery–posterior inferior cerebellar artery (PICA) vessel but no evidence of aneurysm or arteriovenous malformation. Repeat angiography 12 days later revealed a right distal PICA 3-mm aneurysm (Fig. 2B-C). The patient underwent PICA aneurysm clipping (Fig. 3A). She was neurologically intact 3 weeks after the initial Chiari decompression surgery and at 18-month follow-up (Fig. 3B).

Discussion

Normally, pressure equilibrium between the two sides of an aneurysm wall prevent aneurysm rupture.[4,5] Therefore, factors that *rapidly* increase intra-aneurysmal pressure or decrease the pressure around the aneurysmal wall can lead to aneurysm rupture.[4] Decompression of type 1 Chiari malformations can rapidly resolve abnormal CSF flow hydrodynamics, affecting the pressure equilibrium between the sides of the aneurysm wall significantly.[1,2]

In our case, SAH occurred during bony decompression before dural opening. One plausible explanation is that the significant CSF flow changes at the foramen magnum decreased the CSF pressure against the outside aneurysm wall to such an extent that the intra-aneurysmal pressure rapidly and significantly exceeded the inward pressure from outside the aneurysm.

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Two previous cases of remote unknown aneurysm rupture have been reported. A remote unknown cerebral anterior communicating artery aneurysm ruptured during transsphenoidal resection of a pituitary adenoma. After tumor removal, the arachnoid abruptly collapsed into the sella turcica, applying a traction force on the wall of the unknown aneurysm similar to that which may have been created by the sudden large increase in caudal CSF flow in our case. Unlike our case, however, the authors could not exclude intraoperative trauma as a cause of aneurysm formation.[3] The other case coincided with subdural drain insertion after urgent evacuation of a chronic subdural hematoma via frontoparietal craniotomy. CSF drainage may have affected the CSF flow dynamics and aneurysm pressure equilibrium enough to cause rupture.[4] Although our case did not involve draining excessive CSF, the decompression may have caused excessive caudal flow of CSF to the spinal region, which may have precipitated aneurysm rupture.

This is the first report of intraoperative rupture of an unknown cerebral aneurysm immediately after dural opening during Chiari decompression. Only one prior unknown cerebral aneurysm rupture without the possibility of surgical trauma has been noted and that case also occurred when CSF hydrodynamics were affected by CSF drainage with a subdural drain. The significant and rapid changes of previously abnormal CSF physiology noted with decompression of Chiari type 1 malformation can provide a pressure gradient that may predispose to rupture of unknown cerebral aneurysms

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Figure 1. Midsagittal T1-weighted MRI without contrast showing caudal descent of cerebellar tonsils below foramen magnum.

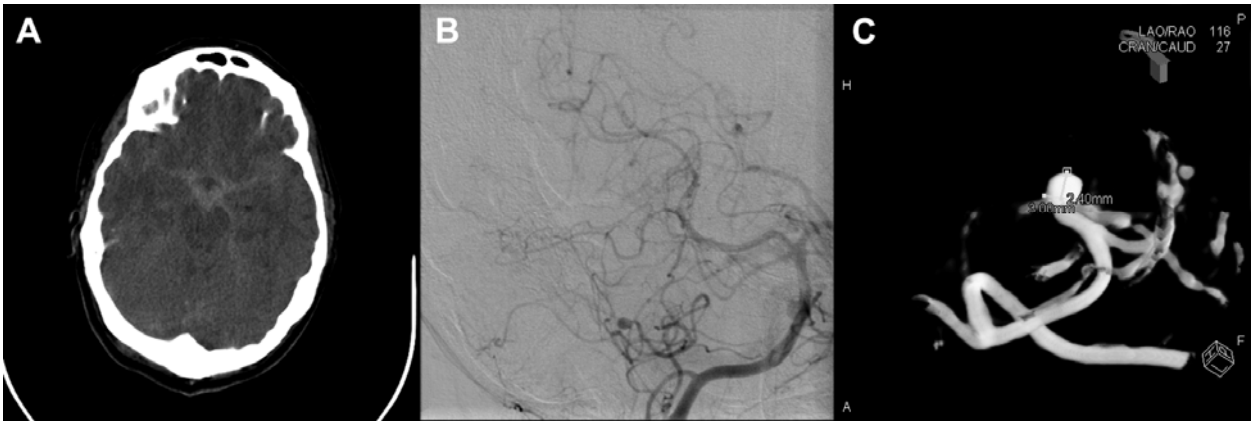

Figure 2. A. Axial CT showing blood in the basal cisterns consistent with subarachnoid hemorrhage. B. Posterior-anterior conventional cerebral angiogram, right vertebral injection showing right-sided vermian distal PICA aneurysm. C. Three-dimensional angiogram showing the aneurysm measuring about 3×2.4 mm in its largest diameters.

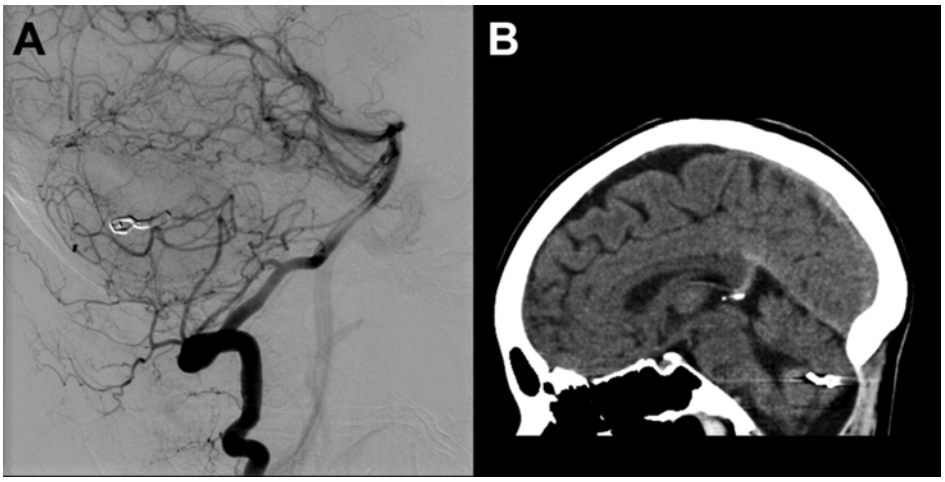

Figure 3. A Lateral conventional cerebral angiogram, right vertebral artery injection, showing complete obliteration of the right PICA aneurysm after aneurysm clipping. B. Sagittal CT scan without contrast obtained 18 months after the patient's discharge showing sufficient decompression at the foramen magnum and stable appearance of the right distal posterior inferior cerebellar artery clip.